Pediatric Bladder Augmentation in Developing Country: Lessons Learnt From an Experience of 195 Cases

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***Abstract**: We report our experience with 195 children who had undergone bladder augmentation in our department during the period 1997 - 2009. These included 78 children with neurogenic bladder, 73 children with incontinent exstrophy and 27 children with posterior urethral valves. While incontinence was a major clinical symptom in many of these children, 107 children had hydroureteronephrosis and 53 had elevated levels of serum creatinine preoperatively. Tissue used for augmentation includes sigmoid colon in 118, ileocecal segment in 39, ileum in 21 and a dilated ureter in 14. Concomitant surgery included bladder neck procedures (Young Dees Leadbetter plasty, bladder neck closure, fascial slings) in 80 children, a catheterizable abdominal stoma in 184, and variety of anti reflux procedures. Special techniques of augmentation such as the preperitoneal augmentation and the "doughnut" augmentation are also described. Results (mean follow up period of 5 years, longest 15 years) were highly satisfactory as regards cure of incontinence and stabilization of hydroureteronephrosis. Vesicoureteric reflux resolved in 98 of 129 units studied. Renal failure, if severe preoperatively, continued to progress and was the major cause of late mortality.*

INTRODUCTION

The normal bladder can store urine for several hours at pressures as low as 5cm H_2O. Storage pressures of more than 30cm H_2O are associated with progressive renal damage, hydroureteronephrosis (HUN), vesico ureteric reflux (VUR), urinary infection and reflux nephropathy. High bladder pressures are also often accompanied by urinary incontinence, even if the bladder sphincters are competent. This situation is seen in 3 common pediatric conditions – neurogenic bladder (NB), valve bladder and reconstructed bladder exstrophy (BE). In these conditions, the bladder may be inherently small or rendered stiff by collagen and/or detrusor hypertrophy. These bladders often struggle to void against anatomical, functional or iatrogenic outflow obstruction as in posterior urethral valves (PUV), NB and BE respectively. Incipient renal failure with polyuria complicates the problem as the large urine output quickly overwhelms the limited storage capacity of the bladder.

The clinical problem of incontinence due to non-compliant bladder can sometimes be managed by non-surgical methods such as timed voiding, nightly catheter drainage, clean intermittent catheterization (CIC) and pharmacotherapy. There remain a proportion of children who require bladder augmentation (BA) - a procedure by which the capacity of the native bladder is considerably expanded by the addition of a segment of bowel or ureter. We present our experience in 195 children who underwent BA in our institution.

MATERIALS AND METHODS

Study Cohort

A total of 195 children underwent BA during the period 1997-2009. The primary diagnoses were NB (n=78), incontinent BE (n=73), PUV (n=27) and 17 children with a miscellany of conditions including cloaca, urogenital sinus, bilateral single system ureteral ectopia and bladder tumor. Of these, 128 were boys. The mean age at the time of BA was 6.25 years, although the youngest was a newborn. Preoperative evaluation included the assessment of renal function, upper tracts, VUR, bladder capacity and urodynamics when feasible. Elevated serum creatinine was present in all cases of PUV, 21 with NB and 5 with BE. HUN was present in all cases of PUV, 66 of NB and 14 of BE. A clinical, radiological, and urodynamic assessment of bladder outflow competence was also done to enable a decision regarding bladder neck procedure during BA, especially in children with neurogenic bladder. When NB was associated with HUN, it indicated high pressures developing in the bladder. After BA, these pressures would be expected to be drastically reduced and incontinence would be relieved without any bladder-neck surgery. NB without upper tract changes, which comprised only 12 cases were assessed urodynamically for leak point pressures. If this was low (below 20cm H_2O), a bladder-neck procedure was planned along with BA. An evaluation was also made as to whether the native urethra would provide an adequate channel for CIC after BA, especially if a bladder neck procedure was contemplated.

Preoperative counseling of the patient and family was considered essential for the success of BA. Expected benefits of BA including dryness and protection of the upper tracts and the disadvantages of BA such as changes in life-style were explained to parents. Life-style changes necessitated by BA included CIC, the likely presence of a small abdominal stoma, bladder washouts and lifelong follow up for potential complications.

The operative procedure

The standard requirements of BA include the choice of a hollow viscus segment of adequate size, which should also be de-tabularized before attachment to the bladder. The bladder should be opened wide to receive the augmentation. Technical details of standard BA procedure have been described elsewhere[1].

In the 195 children, we have used sigmoid colon (n=118) ileocecal

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region (n=39) ileum (n=21) and dilated ureter (n=14) to augment the bladder; 2 cases underwent composite BA and 1 child had auto-augmentation. The ileocecal segment with the *'in situ'* appendix was very useful in augmenting small exstrophy bladder ('doughnut augmentation')[2]. In 43 children, the BA was placed in the preperitoneal space (preperitoneal augmentation), while in the rest the BA was placed in the traditional intraperitoneal space.

Concomitant bladder neck procedures were performed in all 73 BE cases in the form of a Young-Dees-Leadbetter bladder neck plasty in 49 and bladder neck closure in 24 cases. Five children with failed Young Dees bladder neck plasty were re-operated and the bladder neck was closed. Bladder neck procedure was also performed in 7 NB (3 fascial sling and 4 bladder neck closure). Children with PUV and 71 children with NB did not have any bladder neck procedure. Concomitant catheterizable stoma (n=184) was provided in all cases of BE and PUV; and in 67 cases of NB. Appendicular Mitrofanoff procedure was preferred in most of them, while in a few a spare ureteric stump after trans uretero-ureterostomy (TUU) has been used for CIC.

Concomitant antireflux surgery

A variety of antireflux measures were used namely augmentation alone, augmentation with reimplantation, and refluxing-to-non-refluxing trans-uretero-ureterostomy (TUU). Ureterocystoplasty was also used as a method of eliminating VUR into the ipsilateral ureter. The results of various methods employed in the context of the primary disease are presented below for 129 refluxing ureters.

The preperitoneal BA

Pre-peritoneal BA (n=43) was an attempt to reduce the likelihood of intra pperitoncal eritoneal urinary leakage from the augmented bladder which is a rare but life-threatening complication of BA. The procedure involved dissecting the peritoneum off the anterior abdominal wall in the lower abdomen, pelvis and from the dome and posterior wall of the bladder to obtain a large preperitoneal space. The peritoneum was then opened cephalad to the bladder and the chosen bowel segment was isolated with the vascular pedicle. Bowel continuity was restored and the bowel segment for BA was brought out via a limited peritoneal opening (fig.1). A standard enterocystoplasty was then performed. This left most of the augmented bladder in the preperitoneal space created in the earlier part of the operation. In our earlier operations of this type, we only closed the skin leaving the linea alba unclosed to allow free expansion of the BA. Since this resulted in a large hernia, we now close the linea alba in front of the BA with no ill effects. This method is not suitable for BE cases as the anterior abdominal wall is scarred from previous surgery.

The "doughnut augmentation"

This modification of ileocecal augmentation (n=23) was devised for small scarred bladders, especially in BE. After the Young Dees Leadbetter bladder neck plasty, the available bladder surface is much reduced and bilateral ureteric reimplantation and implantation of an appendicular Mitrofanoff port can be difficult in the small residual bladder cavity. The isolated ileocecal segment in a "ring" or "doughnut" configuration had been used so that the "in situ" appendix can be brought out as the Mitrofanoff port from within the ring through a hole made in the ileal mesentery (fig.2). So also the ureter, detached from the bladder plate could be brought inside the ring through openings in the ileal mesentery. The ring of bowel was then opened along the anti-mesenteric aspect except a small length of ileum underlying the appendix as it exited from the abdomen to form the catheterizable port. Suturing of inner wall and outer wall to respective sides yielded a de-tabularized augmentation. As the inner edges were being sutured to one-another, the ends of the ureters were anastomosed to this inner bowel edge, thus effecting the uretero-augment anastomosis. As the outer edges were being sutured, the caudal margin of the opened ileocecal junction was sutured onto the opened bladder, thus effecting the augment bladder anastomosis. At the cephalad end of the completed augment, the small portion of unopened ileum acts as anti-refluxing wrap around the appendicular stoma, thus preventing leakage of Mitrofanoff port. The completed augment included the two ureters within itself, thus providing a very effective mechanism to prevent VUR.

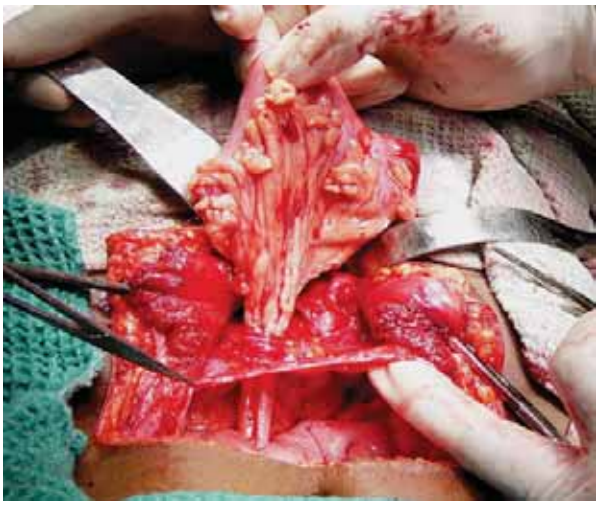

***Figure 1**: Preperitoneal BA: The bowel segment has been brought into the preperitoneal space with its pedicle passing through a limited peritoneal opening.*

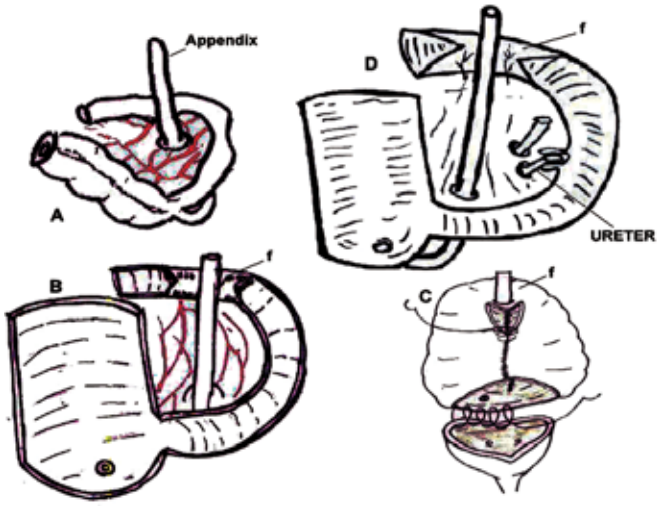

***Figure 2**: Doughnut augmentation: The ileocecal segment has been arranged in a ring like configuration, with the appendix brought into the ring from behind the ileum and to exit from the abdomen in front of the ileum to form the Mitrofanoff stoma (A). The ileocecal segment is opened in the antemesenteric aspect except for a small part of ileum (f) behind the appendix (B). Suturing of 'inner wall'-to-'inner wall' and 'outer wall'-to-'outer wall' yields a de-tabularized augmentation, the lower part of which is attached to the bladder. The segment f acts as antireflux wrap around the appendix (C). If the ureters require reimplantation into the BA, these are brought into the ring and anastomosed to the inner ileal edge (D).*

RESULTS

Mortality

There were 13 deaths in 195 children; 2 deaths were caused by postoperative sepsis-one child was in chronic renal failure and the other had chemotherapy for a bladder tumor. Nine children died due to progressive renal failure several years after BA. Two other children died at home, possibly due to intraperitoneal urinary leak.

Follow up

157 children have been followed up for a mean of 5 years. 25 children are lost to follow up. Resolution of incontinence had been achieved in all valve bladders, in all NB and in 66 BE children. However, this necessitated closure of the bladder neck in 33 children. The Mitrofanoff channels had also remained continent unless CIC was unduly delayed. One Mitrofanoff channel had to be closed for incontinence, while five others needed a skin level revision for stenosis.

The upper tracts which were normal prior to BA, remained without deterioration; those with HUN showed resolution or stabilization of calyceal dilatation in all but 3% of cases. Renal failure, when present preoperatively showed improvement or stabilization only when the serum creatinine was within twice the expected maximum for age. This was the case in most NB and EB. In children with PUV, those with preoperative creatinine levels more than 2 mg/dl did not show stabilization in the period of follow up[3]. Eight of the 9 children dying from renal failure within the follow up period were of the PUV group. One child augmented for NB in renal failure, stabilized till she underwent renal transplantation as an adult.

Resolution of VUR showed a varied pattern dependent on the primary condition as well as the anti-reflux procedure undertaken. Post-operative cystograms were not routinely done; they were available for 129 renal units which had been refluxing prior to BA. Overall, 98 of these (76%) had ceased to reflux. Augmentation alone, without any other specific anti reflux surgery resulted in resolution of VUR in 71% of 14 units in NB, 100% of 8 units in PUV and in only 40% of 20 units in BE. Thus, VUR resolved in 26 of 42 refluxing units by augmentation alone (Fig. 3,4). Seventy three refluxing units underwent ureteric reimplantation along with BA. VUR resolution was seen in 86% of 21 units in NB, 100% of 2 units in PUV, 79% of 42 units in BE when a reimplant was done into the bladder segment and 63% of 8 units in BE when the ureter was implanted into the bowel segment (This excludes the ureters implanted into the doughnut augment). When a contralateral non-refluxing but dilated ureter was available, refluxing-to-non-refluxing TUU in 13 ureters, (8 in NB and 5 in PUV) resulted in 100% reflux resolution. The refluxing ureteric stump thus available was often used as a catheterizable port as the augmentation often reduced the reflux into the stump to a manageable level.

COMPLICATIONS

Apart from complications attendant on any major abdominal surgery such as adhesive intestinal obstruction, wound dehiscence, anastomotic leak, the life threatening complication specific to BA was intra-abdominal urinary leak (5 of 195 cases with 2 deaths) These leaks were seen only in the traditional intra-peritoneal type of augmentation (n=152) and not in the 43 children with the preperitoneal BA. Of three of these children who reached the hospital with intra abdominal leak, one required re-laparotomy, while 2 settled with bladder drainage. Calculi were seen in the augmented bladder in 5 children, 4 of whom were BE cases wherein the bladder neck had been closed. The calculi were surgically removed. Metabolic complications of osteodystrophy, hypokalemia and acidosis were seen in 9 children, all of whom were in chronic renal failure.

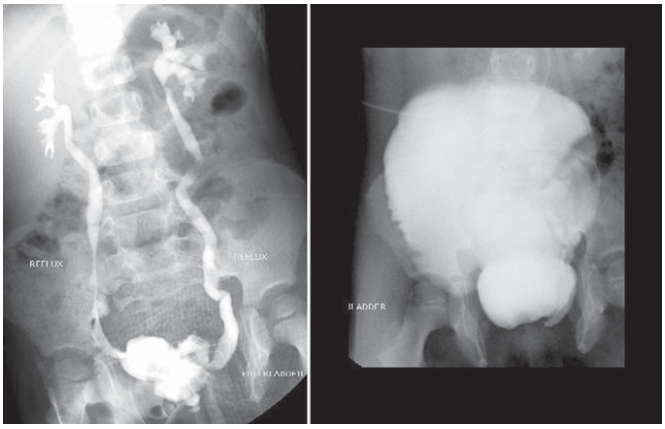

Figure 3: *The benefits of augmentation: Voiding cystourethrogram in a case of incontinent exstrophy with a small bladder and VUR (A); after BA and bladder neck plasty (B) has relief of incontinence and resolution of VUR.*

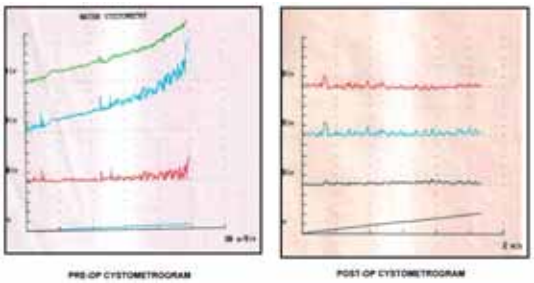

Figure 4.: *Urodynamic graph showing drastic reduction of bladder pressure after augmentation.*

DISCUSSION

The most distressing complaint in the majority of these 195 children was urinary incontinence which made them social outcastes, mostly unable to attend school. BA, with or without the aid of bladder neck procedure, made these children dry and this was a tremendous boost in the psychosocial life of these children and their families[4,5]. In addition, BA results in long term benefit in terms of resolution or stabilization of upper tract pathology unless renal failure is advanced. BA, however, incurs necessary changes in life style, such as need for CIC and bladder washouts, and also has inherent long term complications. Thus, the place of BA in pediatric reconstructive urology needs a thorough understanding of the common conditions where BA might be indicated.

BA IN NEUROGENIC BLADDER

Most NB are treated non-operatively and the 78 children who had undergone BA in this series comprise just 26% of the 300 children admitted for neurogenic bladder management in the same period. In 71 of 78 cases of NB, BA was done for a poorly compliant bladder associated with upper tract changes in 66 of these[6,7]. The other 7 cases underwent BA for intractable incontinence with normal upper tracts. Bladder neck surgery to tighten or even close the bladder neck

was performed with BA. We believe that BA is the safer option in this situation rather than allow a high pressure bladder to develop subsequent to bladder neck surgery without BA, though the literature is polarized on this issue[8,9]. When CIC becomes universally practiced in the management of NB, the need for BA might decrease, though this remains to be proven[10].

BA IN EXSTROPHY BLADDER

In our experience, reliable dryness in repaired exstrophy can be obtained by BA along with bladder-neck plasty and that BA will be required in the majority of children with exstrophy[11]. In this series, 73 children underwent BA, while 99 children were treated for exstrophy in the same period, thus BA being done in 74% of cases. Eight of 10 children who achieved dryness after exstrophy reconstruction without BA, gradually developed incontinence and/or upper tract changes as late as 10-15 years after reconstruction and underwent BA at that time. This experience corroborates with the long term results of BE reported in the literature[12-14].

BA IN POSTERIOR URETHRAL VALVES

While the primary management is valve fulguration, the diseased bladder needs careful and continued management. BA has a place when conservative treatment has failed to resolve a poorly compliant, high-pressure bladder causing progressive upper tract deterioration and incontinence. However, since most valve bladders improve with time and become compliant, the decision to augment requires considerable judgment. The 27 children who had undergone BA for posterior urethral valve disease comprise 8% of 334 children treated for this condition in the same period.

CHOICE OF HOLLOW VISCUS FOR BA

The most common bowel segment chosen for BA is the ileum, as it yields a low pressure adequate augment with low malignancy potential. We have, however, chosen the sigmoid colon in most children because of the proximity of this viscus to the bladder and the ease of BA with minimum disturbance to the rest of the abdomen. The sigmoid colon, however, has a higher malignancy potential and can sometimes generate high pressures in spite of de-tabularization. The ileocecal segment has the attraction of an "in situ" appendix for the catheterizable port and we have found it very useful for exstrophy cases with small bladders as a "doughnut augmentation". The ureter has to be adequately dilated for it to be of use in BA and thus it is only occasionally available. The ureterocystoplasty avoids problems of mucus production, metabolic disturbance and malignancy risk[15,16]. Gastrocystoplasty is said to be useful when the child is in renal failure with metabolic acidosis because acid excretion by the stomach patch reduces metabolic acidosis. However, gastrocystoplasty has other major problems such as the hematuria-dysuria syndrome and it has not become a popular method of BA. Experience with de-mucosalized bowel segments and auto augmentation are limited and the only child with auto-augmentation in this series was not benefitted significantly. Bioengineered bladders are experimental. In most situations, enterocystoplasty remains the gold standard for BA[17-19].

Apart from the long-term merits and demerits of various visceral segments used for BA, there are other more immediate concerns to be addressed in the choice of BA. In children with anorectal malformation, for example, ileum could be the best option because of abnormal colonic anatomy or blood supply. Thus colon or ileocolic segment are not to be used. In the child with meningomyelocoele, sigmoid colon preferred over ileocecal segment, as it is often very capacious in these children due to constipation while loss of the ileocecal valve would be detrimental to fecal continence.

CONCOMITANT SURGICAL PROCEDURES WITH BA

The three important issues are bladder neck procedure for continence, provision of a continent catheterizable channel and surgery for VUR. In our experience, a bladder neck plasty is required in BE and a fascial sling for NB with normal upper tract and a low leak point pressure. Children with HUN generally do not need bladder neck procedures as BA alone will cure or drastically reduce the incontinence. We do not prefer to close the bladder neck unless this option is unavoidable as in very small bladder with a widely open neck or previous failed bladder neck surgery. Bladder neck closure can be technically challenging and a rectus abdominis muscle flap interposition is useful[20-23].

CIC and daily bladder washouts are essential after BA, and most of our children have had a continent catheterizable port concomitantly with BA. This is essential when a bladder neck surgery has been done as in BE and preferable when urethral CIC is likely to be difficult or painful as in posterior urethral valves and anorectal malformation. The appendix has been used in almost all children for CIC, while in a minority of subjects a stump of ureter left after TUU was used[24]. When these are not available a tube of ileum (Young-Monti) or greater curvature of stomach was used. We have used the easily available 8 Fr feeding tube for CIC.

The issue of anti reflux surgery concomitant to BA remains controversial[25-27]. It has been reported that BA alone is an effective anti reflux measure. Our results show that this is certainly not true in BE, where anti-reflux surgery is desirable along with BA. In NB and PUV, BA alone is often effective in resolving VUR but not always so. Thus, if a reimplantation can be done safely, it should be done. Reimplantation in the exstrophy bladder caused resolution of VUR in nearly 80% while BA alone was associated with only 40% resolution of VUR. Results of reimplantation in the doughnut augmentation are yet to be analyzed as these have been done mostly after 2009, but our initial results are highly encouraging. Refluxing-to-non-refluxing TUU was a very successful and safe method of VUR resolution if a contralateral non-refluxing but dilated ureter was available.

COMPLICATIONS OF BA

Intraperitoneal urinary leak is a rare, but life threatening complication after BA and it can occur at any time[28-30]. To prevent this complication, we have elected to do the pre-peritoneal BA whenever possible and we have not had any urinary leaks from this procedure. However, the pre-peritoneal BA is not feasible in BE and in children who have had previous lower abdominal surgery. The incidence of calculi in this series has been very low although possibly more such will come to light with longer follow up. Daily irrigation of the bladder via the catheterizable stoma is emphasized and this may be a factor in preventing calculi[31]. A closed bladder neck increases the chances of calculus formation as bladder wash from above may not remove mucus and gravel that collects at the dependant part of the bladder. The majority of children who developed calculi in the BA had a surgically closed bladder neck. Previous history of urinary calculi is also a predisposing factor. Metabolic complications were seen only in children with pre-existing renal failure and thus BA can be difficult to manage in the presence of advanced renal failure, especially in a

developing country. We have advised oral bicarbonate supplementation in all BA cases to avoid latent alkali deficit in the long term. Whenever, ileum or ileocecal segments were used, parenteral vitamin B_{12} was given prophylactically once in every three months[32, 33].

The augmented bladder has an increased risk of malignancy in the long term. We have not encountered this problem as our longest follow up is only for 15 years. In the literature, the incidence is reported to be 7 out of 153 cases after a mean of 32 years following BA[34]. The risk is largely influenced by the inherent malignancy potential of the bowel segment used (stomach and colon have higher potential than ileum or ureter) and of the diseased bladder (exstrophy bladder is more prone to malignancy) Carcinogenic stimuli such as smoking and immune suppression after renal transplantation are additional factors. Thus the onset of malignancy is multi factorial and is not solely due to BA.

CONCLUSION

BA is feasible in a developing country if the family is motivated. When done for the correct indication with meticulous preoperative assessment and postoperative care, BA can make a great improvement in the quality of life and provide long term renal protection.

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